

Practical Considerations for Using Online Methods to Engage Patients in Guideline Development

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Abstract Clinical practice guidelines (CPGs) have been widely used in healthcare policy, practice, and for suggesting future research. As patients increasingly become involved in CPG development to produce patient-centered recommendations, more research is needed on methods to engage patients, particularly methods allowing for scalable engagement of large, diverse, and geographically distributed groups of patients. In this article, we discuss practical considerations for using online methods to engage patients in CPG development. To inform this discussion, we conducted a rapid, systematic review of literature on patient involvement in CPG development and used qualitative evidence synthesis techniques to make inferences about potential advantages and challenges of using online methods to engage patients in this context. We identified 79 articles containing information about involving patients in CPG development. Potential advantages include the ability of online methods to facilitate greater openness and honesty by patients,

as well as to reflect the diversity of patient views, which in turn further improve the utility of CPGs. Potential challenges of using online methods may include the extra skill, time, and certain types of resources that may be needed for patient engagement, as well as the difficulty engaging specific patient populations. However, these challenges are mitigated by growing calls for patient engagement as normative for CPG development in addition to patients' increasing familiarity with online technologies. These practical considerations should be examined empirically as guideline development groups further explore the appropriateness of using online methods to engage patients across different stages of CPG development.

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Key Points for Decision Makers

Potential advantages include the ability of online methods to facilitate greater openness and honesty by patients, as well as reflect the diversity of patient views, which in turn further improve the utility of clinical practice guidelines.

Potential challenges of online methods include the extra skill, time, and certain types of resources that may be needed for patient engagement, as well as the difficulty engaging specific patient populations.

Online engagement is most likely to be useful when patients seek anonymity to feel comfortable to share their views openly; a large, diverse, and geographically dispersed group of patients is sought; patients have access to and proficiency with online technology; and the guideline development group has time and resources to train patients, ensure they understand their tasks, and manage the needs of special patient populations.

1 Introduction

1.1 Expanded Use of Clinical Practice Guidelines (CPGs)

Clinical practice guidelines (CPGs) have been widely used in healthcare policy, practice, and research [1]. Originally intended to support clinical decision making by individual clinicians, CPGs now also inform institutional policies, insurance coverage, quality measures, and legal liability [2]. Moreover, the number of CPGs and organizations creating them have proliferated exponentially over the last several decades, with thousands of CPGs archived in the International Guideline Library of the Guideline International Network (G-I-N) alone [3]. CPGs are receiving significant attention by healthcare stakeholders seeking to ensure the trustworthiness of CPG recommendations [1]. In addition to the quality of CPGs, researchers and clinicians want to know whether CPGs facilitate shared decision making between clinicians and patients, patient-centeredness of CPGs improves their relevance to patients' experiences, and the quality of the actual clinical decisions made [4].

In this article, we discuss practical considerations for using online methods to engage patients in CPG development based on a rapid review of existing literature on patient engagement in the CPG context. We focus on online methods because of their potential for scalable engagement of large, representative, and geographically distributed patients.

1.2 Patient Involvement in Guideline Development

Patient involvement in CPG development is increasingly common and can take various forms [5, 6]. The G-I-N Patient and Public Involvement Working Group (G-I-N PUBLIC) continually encourages international collaborations to strengthen existing knowledge, development, and evaluation of patient and public involvement in CPGs [7]. The UK National Institute for Health and Care Excellence (NICE) has involved patients and the public in the development of CPGs for the UK National Health System to increase the relevance of CPGs to patients most directly affected by their recommendations [8]. The World Health Organization (WHO) includes patients as part of guideline development groups (GDGs) and solicits their input on the key questions to be answered in the CPG [9]. Numerous societies such as the American College of Physicians (ACP) and the American Academy of Neurology (AAN) involve patients in CPG development to varying degrees [10, 11]. The US Institute of Medicine (IOM) also recommends that, for CPGs to be trustworthy, GDGs should

include a current or former patient and a patient advocate, as well as involve patients in reviewing draft CPGs prior to their finalization [1].

The increased involvement of patients in CPG development has led to significantly more attention paid to the specific modes and methods for engaging and including patients [12]. Patients and patient organizations tend to favor inclusion of patients in CPG development so that their unique knowledge, experience, and perspectives have the optimal chance of adequately being captured in guideline recommendations [13, 14]. With some exceptions, involvement of patients typically means including only a few patient representatives on GDGs rather than proactively engaging a wider group of patients in the development of recommendations due to budgetary and logistical constraints [1, 9]. Online methods are therefore a promising modality for systematic and large-scale patient engagement in CPG development.

1.3 Online Methods of Engaging Patients

Leaders of GDGs often need effective yet efficient methods for engaging patients in the CPG development process, making in-person engagement methods challenging for more than a small number of patient representatives [4]. Patients have found proactive methods of online engagement to be acceptable for other healthcare areas, such as research prioritization and performance measure development [15, 16]. However, less is known about the applicability of systematic and scalable methods for engaging patients online in the area of CPGs, as well as the practicalities of expanding patient engagement across the various stages of CPG development.

2 Methods

2.1 Context: Project Developing an Online Method for Engaging Patients in CPG Development

We are members of a project team developing a scalable, online method for patient and caregiver engagement in CPG development [17], modeled after an approach to soliciting clinicians' input in the same process [18]. A specific aim of this project is to create a protocol that patients with Duchenne muscular dystrophy—a rare, progressive disorder with pediatric onset—and their caregivers can use to rate the perceived level of patient-centeredness of CPG recommendations, using the ExpertLens system [19]. ExpertLens is an online modified-Delphi system [15, 16] that facilitates the administration of expert panels conducted using the RAND/UCLA Appropriateness Method for soliciting clinician input in CPG development [18]. A key aspect of developing this protocol has involved

the practical considerations for using an online engagement method when patients and caregivers are geographically dispersed and, in our specific context, with patients who have mobility challenges that make travel to in-person meetings difficult, impractical, or even impossible.

2.2 Design: Rapid Review Using Qualitative Evidence Synthesis Methods

To inform practical considerations of a scalable, online method for patient engagement in CPG development, we combined two innovative research synthesis methods—rapid review and qualitative evidence synthesis methods [20]—to review relevant literature on patient involvement in CPG development. Our review focused specifically on the types of patients typically involved in CPG development, the stages in which patients were engaged, the methods used to engage patients, and the results of patient engagement. As with systematic reviews, a rapid evidence synthesis or ‘rapid review’ relies on explicit, systematic methods for identifying, selecting, extracting, and summarizing findings from the research literature. In contrast to full systematic reviews, they involve concessions in the breadth and depth of these processes to meet specific informational needs in a shorter timeframe [21] and are often used to explore and organize themes in an emerging research area [22]. We conducted a rapid review rather than a systematic review to meet our project goals and demands within contractually approved timeline and budget [23]. Recent studies suggest that rapid reviews are useful for translating knowledge from research into practice to ensure timely development of evidence-based decisions [24]. We analyzed information extracted from the identified literature using qualitative evidence synthesis techniques to make inferences about potential advantages and challenges of using *online* methods specifically for engaging patients in CPG development. As with quantitative evidence synthesis (i.e. meta-analysis), qualitative evidence synthesis involves extracting and combining evidence from individual studies addressing the same topic, although does so by categorizing and summarizing descriptive *textual* rather than numerical data [25]. We used reporting standards for qualitative research [26, 27] and systematic reviews [28] to prepare this report.

2.2.1 Search Strategy

In collaboration with research librarians, we conducted a literature search of four electronic databases of peer-reviewed literature (Cumulative Index to Nursing and Allied Health Literature [CINAHL], PsycINFO, PubMed, and

Web of Science) and one database of grey literature (New York Academy of Medicine) from inception to 20 October 2016. Search strings included terms related to ‘guidelines’, ‘patients’, and ‘patient centeredness’. We also assessed citations suggested by the study team and advisory board.

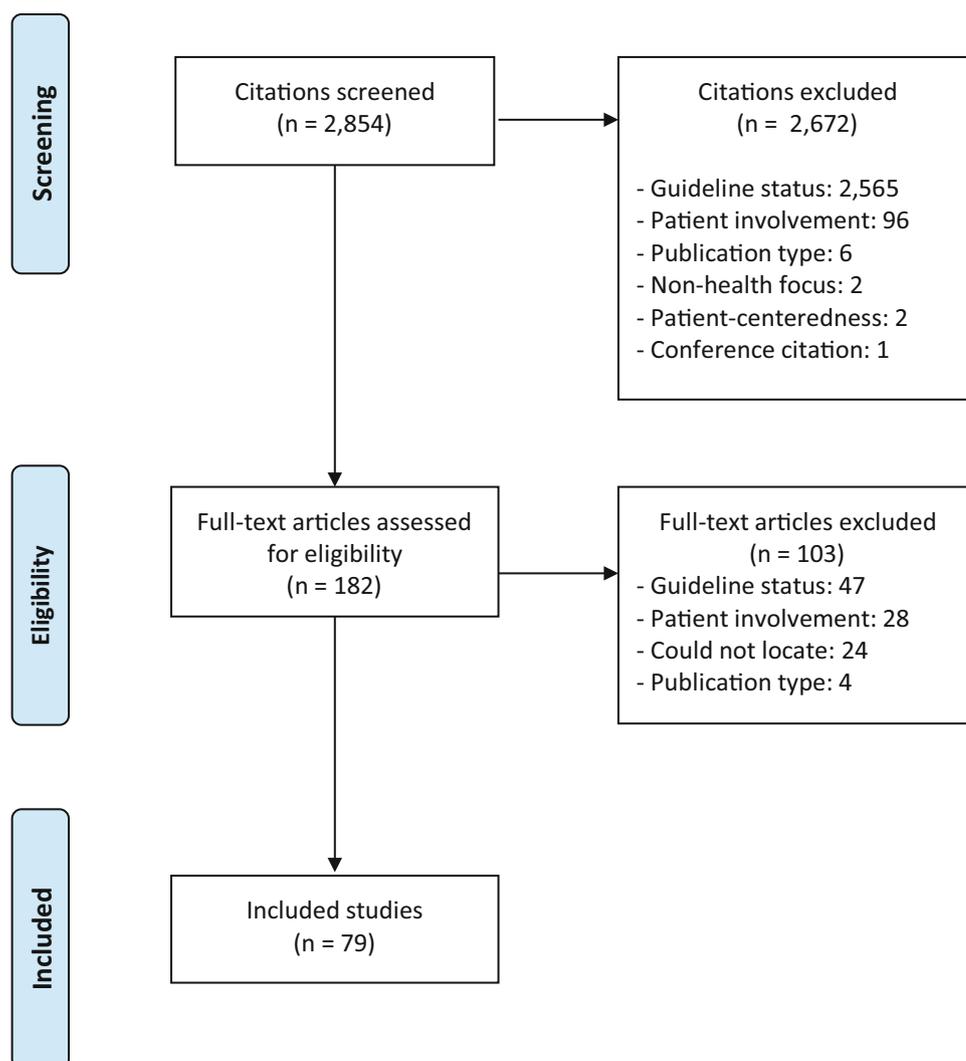
2.2.2 Eligibility Screening and Assessment

We included articles published in English that discussed patient involvement in CPGs, and excluded conference citations, newspaper and magazine articles, dissertations and theses, and books and book chapters. Articles that did not discuss CPGs or that discussed CPGs but did not mention involving patients or addressing patient-centeredness were also excluded. Because we were interested in conceptual discussions of and empirical findings on patient involvement in CPG development, and because we did not expect to find many articles on online engagement specifically, we did not set any eligibility criteria related to type of study or mode of patient engagement. As is typical for rapid reviews [21], one researcher (SG) screened all retrieved citations according to these eligibility criteria and then assessed all full-text articles judged as potentially eligible at the citation screening stage.

2.2.3 Data Extraction and Synthesis

Through iterative team meetings and a presentation to our project’s advisory board, we developed a data extraction list for the rapid review. With consultation from the study team, one researcher (SG) extracted the following information from each included article: type of study, geographic area, clinical topic, who represented the patient perspective during CPG development, stages of CPG development that involved patients, how patients were involved in CPG development, and the reported impact of including patients in CPG development. This researcher then used a qualitative evidence synthesis technique known as thematic synthesis to make inferences about the practicalities of online methods for patient engagement in this context [29]. This synthesis involved three stages: line-by-line coding of relevant text, development of descriptive themes that speak directly to the line-by-line coded data, and inference of more abstract analytical themes from descriptive themes related to potential advantages and challenges of using online patient engagement methods. We used a 10-step framework outlining steps and options for continuous patient engagement in CPG development to guide our analysis [12]. The strategy for and findings from the thematic synthesis were presented to the project team and revised in an iterative fashion until all project team members’ comments were addressed.

Fig. 1 Flow of studies through the literature search



3 Results

3.1 Results of the Search

We identified 2854 citations through our search, of which we considered 182 citations as potentially of interest based on the information reported in the titles and abstracts. Of these 182 citations, we excluded 103 articles, primarily because they either did not focus on guidelines ($n = 47$) or did not report involving patients ($n = 28$). Overall, we reviewed 79 articles (Fig. 1).

3.2 Characteristics of Reviewed Articles

We identified articles covering various study designs, clinical topics, and geographic areas (Table 1). The most common study designs were conceptual articles and

literature reviews (22% each). Most (49%) specified a physical health condition, such as cancer, chronic obstructive pulmonary disease, or rheumatologic issues, or health in general (43%), although some articles were focused on mental health conditions (8%). The majority of studies (66%) were national in scope, most often the US, UK, Canada, The Netherlands, or Australia, although a substantial proportion had a global remit (23%). The remaining articles (11%) focused on a specific continent (either North America or Europe). The majority of studies discussed representation of the patient perspective by patients themselves (90%) and patient engagement during the CPG stage of developing recommendations (84%). Overall, 21 (27%) articles explicitly discussed an online method of engagement, including online public comments, Delphi processes, voting tools, and Wikis or discussion forums.

Table 1 Characteristics of reviewed studies

Variable	N (%)
Study type	
Conceptual article	17 (22)
Qualitative primary study	15 (19)
Quantitative primary study	7 (9)
Formal consensus method	7 (9)
Literature review	17 (22)
Clinical practice guideline	10 (13)
Methods manual for guidelines	6 (8)
Clinical topics	
Health generally	34 (43)
Specific physical health condition	39 (49)
Specific mental health condition	6 (8)
Geographic area	
National	52 (66)
Continental	9 (11)
Global	18 (23)
Discussed online engagement	
Yes	21 (27)
No	58 (73)
Representative of patient perspective	
Patients themselves	71 (90)
Caregivers/family members	18 (23)
Representatives/advocates/consultants	32 (41)
Healthcare consumers	22 (28)
The ‘public’ generally	19 (24)
Guideline development stage	
Nominating guideline topics	8 (10)
Prioritizing guideline topics	11 (14)
Selecting guideline group members	5 (6)
Framing the question	25 (32)
Creating a research plan	7 (9)
Conducting a systematic review	9 (11)
Developing recommendations	66 (84)
Dissemination and implementation	19 (24)
Updating	2 (3)
Evaluating methods and impact of engagement	21 (27)

3.3 Practical Considerations for Online Patient Engagement

We identified several advantages, challenges, and mitigating factors of using online methods to engage patients in CPG development (Table 2). In terms of advantages, online methods can facilitate greater openness and honesty by patients than in-person methods, in which patients can be resistant or uncomfortable with more direct engagement [12]. Clinicians may dominate in-person meetings,

potentially leaving patients frustrated with a limited focus on the patient experience [30], or deferential to the opinions of professionals at the expense of their actual views [31]. Through their ability to assure anonymity, online methods can protect against social and psychological pressures common among in-person methods. For instance, online Delphi processes can protect patients against group influences and the authority of professionals, and allow them to suggest “their opinions and proposals might be more realistic” [32]. In addition, online methods can facilitate patient engagement that reflects the full diversity of patient views. With online methods, GDGs may find it easier to increase the number of patients engaged [33], recruit statistically representative samples of patients [34], ensure greater diversity of patient opinions [35], include patients with limited mobility or other travel limitations [32], and include patients during different phases of guideline development [12, 36].

Because of the above-described advantages, online methods can further increase the utility of CPGs. Patients often place different values than clinicians on certain issues addressed within a CPG [36], particularly those related to patient education and self-management [37]. Patient engagement can help ensure the CPG content and recommendations adequately reflect their views and preferences [36], improve the readability of patient summaries [38], and increase the chances of a receptive audience upon dissemination [39]. Direct patient input may even result in a number of changes to CPGs themselves [40]. Patients can play a particularly vital role in the implementation of recommendations in clinical practice if numerous patients are proactively included in developing and pilot-testing patient summaries of guidelines [38, 41].

We also identified several potential barriers that may serve as challenges for online methods of patient engagement. First, online methods may require even more skill, time, and certain types of resources to engage patients than in-person methods. GDGs using online methods may experience more difficulty training patients to fully understand their tasks, particularly the more technical language and processes of CPG development related to scientific proficiency [42]. More intensive training, phone and email support, time to complete the engagement task, and/or simplified engagement tasks may be needed to allow for large-scale engagement of patients online [7, 43].

Another challenge is the perception that online methods are less engaging than in-person methods [44], given the importance of genuine (rather than tokenistic) engagement to patients. Patients may feel loss of an important ‘human touch’ when using online methods [40]. Relatedly, online engagement methods can be difficult for specific patient populations. In-person methods are more flexible for exploring and identifying complex sources of diversity in

Table 2 Practical considerations for online patient engagement identified in the literature

Analytic theme	Descriptive theme	Example quotation
Potential advantages		
Facilitates greater openness and honesty	Dominance by clinicians during in-person meetings	“Collective health care decision-making processes like guideline development, health technology assessment, and clinical priority setting are often chaired by content experts and people in a hierarchical position of power (e.g., lead clinician and CEO). Our findings show that group moderation by an expert in group processes (rather than by a content expert) could help even out existing power differences, facilitate more fruitful deliberation, and support professionals’ and public members’ mutual understanding and influence” [59]
	Deference to experts during in-person meetings	“The professional perspective is dominant and patients look up to scientific experts. Therefore, patients’ experiential knowledge and input are easily overruled, perhaps unintentionally. Professional hierarchy (e.g. titles) can be a factor for reluctance or nervousness to speak up during these meetings. It is reported that patients for this reason may adopt the opinion of professionals or change their own views” [31]
	Protection from social/psychological pressures online	“Most patients have difficulties with holding their own when facing a team of professionals; becoming easily overruled by professionals causing the collaboration to degenerate into tokenism. This was observed in fact with patient participation in the Steering Committee responsible for the coordination of the CPG development. However, the Delphi consultation avoided face-to-face interactions between clinicians, researchers and patients, overcoming this barrier and also preventing contamination effects among patients. Participants were therefore protected from the influences of the group and the prestige or power of other contributors, suggesting that their opinions and proposals might be more realistic” [32]
Greater ability to reflect diversity of patient views	Representative samples	“A statistically representative sample of patients with direct experience of the disease are seen as more reliable and less biased than the views of individual patients’ representatives participating in CPG development” [34]
	Larger samples	“Via WikiFreya, 298 patients formulated 289 recommendations, which 80 patients prioritized into 21 recommendations. These recommendations were included in the definitive guideline. Twenty-one prioritized patients’ recommendations, obtained via Wiki-Freya, were included” [33]
	Diverse samples	“There is variability in patient values and preferences at different stages of disease (new-onset, stable, acute exacerbation, rehabilitation, palliation, and terminal care), with different disease severity (from mostly asymptomatic to critically ill), and when considering different issues (for example, when considering testing, medication choice, surgical treatments, intubation, and whether to enter a hospice). Values and preferences may differ across age and sex, socioeconomic status, ethnicity, and culture” [35]
	Geographically dispersed samples	“The Delphi consultation improved its efficiency by the use of electronic mail This study succeeded in recruiting a significant number of patients from most Spanish regions rapidly and inexpensively through the use of communication technologies” [32]
Improved utility of CPGs	Improved clinical recommendations	“Allow consumer views to either complement the technical knowledge of non-consumer experts or challenge any previously held assumptions, both of which may improve the quality of guideline content” [60]
	Improved implementation	“Consultation with stakeholders during guideline production can enhance uptake because it engenders a sense of ownership by addressing their concerns” [43]

Table 2 continued

Analytic theme	Descriptive theme	Example quotation
Potential challenges		
Compounds the required resources, skill, and time	Less engaging than in-person	“The structured peer-facilitated workshops enabled active discussion about topics and outcomes that were important to the participants. The workshop format helped to facilitate rapport between the participants; and cohesiveness in diversity was observed in which the participants shared a range of experiences and views but worked cohesively as a group... . The peer interaction promoted idea generation as the participants were observed to be building ideas through sharing their experiences, opinions and perspectives, and asking questions of each other. The use of flipcharts promoted a high level of interactive discussion, and the visualization and generation of ideas” [44]
	Required scientific proficiency	“Individual patients who participated in a guideline development group contributed infrequently and had problems with the use of technical language. Although they contributed most in discussions of patient education, their contributions were not subsequently acted on. Within a ‘one off’ meeting with a group of patients, participants again encountered problems with medical terminology and were most interested in sections on patient education and self management. Their understanding of the use of scientific evidence to derive more cost-effective care practices was unclear. The workshop format was relative resource intensive but made it possible to explain the technical elements of guideline development, enabling patients to engage in the process and make relevant suggestions” [42]
	Required training	“Participants pointed towards the importance of recruitment, support and training as key conditions for meaningful involvement of patient and public representatives. Training may cover the fundamentals of guideline development and approaches for reporting back to consumer constituencies, or offer mentoring opportunities from other patient/public representatives” [7]
	Required time	“The process can be costly. Involving stakeholders in decision-making demands commitment from the entire organization, specific managerial arrangements, and sufficient backup. This can prove to be a burden for organizations with insufficient funding. For example, NICE guidelines average 200 registered stake-holder organizations that comment on the scope and drafts. Administering, collating, and responding to these comments takes over 4 weeks of several people’s time, including the guideline group chair. To be successful and trusted, such a process requires commitment from the entire organization, specific managerial arrangements, and adequate resources” [43]
Greater difficulty engaging specific patient populations	Professional patients	“There is debate whether partners who contribute over longer periods of time start to become ‘professional’ patients. Professionals might argue that these partners risk losing touch with their fellow patients and alienate themselves from the target group, or they might represent the views of special interest groups” [31]
	Bias toward younger groups	“Ideally, to test the feasibility of such a new method for patient participation in CPG development, an Internet-using young target group such as infertile patients is preferred. This wiki has been tested in the field of infertility care, representing a relatively young target group. More than 98% of this group use the Internet. This participant characteristic is associated with more frequent health-related Internet use. Therefore, the participants in our study were an ideal subgroup for testing and evaluating a wiki-based method, which argues against the generalizability of our findings to other patient groups” [46]
	Overly-passionate patients	“Another challenge with consumer involvement occurs when the representative has a visceral personal experience with the disease or an advocacy role that interferes with the ability to examine the evidence and recommendations dispassionately. Such individuals may have difficulty divorcing their personal narrative or policy agenda from the systematic methods and analytic rules a practice guideline group should follow” [42]

Table 2 continued

Analytic theme	Descriptive theme	Example quotation
Greater difficulty engaging specific patient populations	Patients with more education	“It must be noted, however, that the participants were all college educated, and that including less well-educated patients would require additional time and effort to prepare the panel members to effectively participate” [47]
	Patients with limited Internet access	“In Australia, internet access and connectivity is still restricted in some rural areas and online consultation presents a participation barrier for stakeholders with limited access. Therefore, we continue to support offline channels until Australian communication infrastructure enables the rural population to participate more actively online” [48]
	Caregivers as proxies	“It is important to recognize that caregivers’ and patients’ interests sometimes conflict. Furthermore, family members, nonprofessional caregivers, and health professionals all share large degrees of inaccuracy in ascertaining or predicting patient wishes or expectations” [35]
	The public as proxy	“Involving the wider general public involves challenges beyond involvement of COPD patients and their caregivers. Public views are often based on hypothetical judgments (compared with patients and caregivers, who can draw on personal experience). Public views on the use of public funds, for instance, may differ from those of people affected by the condition (for example, placing a higher value on life-prolonging treatment vs. improvement in quality of life). The general public may have beliefs that conflict with the interests of patients and caregivers. This may be critically important for guideline panels who may be increasingly expected to take resource use into account. For example, the public may not be aware of the importance of COPD and may be unwilling to recommend resource-intensive treatments for conditions that are perceived as “self-inflicted”: the stigma associated with smoking adversely affects people with lung cancer and COPD. The wider public also includes patients with conditions other than COPD and, therefore, some of them may have competing interests where health service resources are limited” [35]
	Patients with treatment experience	“Whether patients had experienced the treatments under consideration appeared to influence results. Typically, previous exposure with a given treatment was associated with a preference for continuing that same treatment. Cognitive dissonance occurs when participants are inclined to modify their interpretation of information to ensure that it is consistent with their previous decision. To reduce cognitive dissonance, participants who had previously been exposed to the treatments under consideration may be inclined to continue their treatment, even in the face of information suggesting that it is not the optimal choice. Patients who do not want to believe that they have been taking the wrong treatment may interpret the evidence presented so that it is consistent with their prior choice” [45]
	Patients with chronic conditions	“Most initiatives for patient involvement in clinical guideline development have been carried out for chronic diseases. These patient groups are often united in patient organizations, are usually motivated to participate and are therefore relatively easily accessible. The involvement of patients in guideline development with incidental and non-threatening diseases (e.g. hysterectomy, treatment of pneumonia or concussion) is more complicated. These patient groups are most often not united in patient organizations, and patients are only ‘patient’ for a limited period of time. As a consequence, the inclusion of patient representatives in a guideline workgroup cannot easily be realized and is less appropriate because patients lack the broader input from the collective knowledge of the patient organization and the experiences between individual patients differ greatly. Moreover, after recovery, patients most often want to forget their (negative) disease experiences and want to continue with their life. Little knowledge is available on how patients with incidental and non-threatening diseases can most effectively be involved in clinical guideline development” [49]

Table 2 continued

Analytic theme	Descriptive theme	Example quotation
Mitigating factors		
Increasing internet use	Familiarity with online methods	“There is increasing evidence of the applicability of electronic communication platforms for the population with TBI. Indeed, approximately one person in two with TBI has access to the Internet and around 60% of them use Facebook on a regular basis” [50]
	Proactive use of Internet for healthcare	“Historically, patients were told what their treatment would encompass through a one-directional flow of information. However, with the internet as an emerging source of information, patients increasingly ‘Google’ and ‘Wiki’ their diagnosis and can be more informed than their clinical team” [38]
	Demonstrated online recruitment	“For the patient panel, infertile patients were recruited through a call for participation at ‘Freyawiki’ (the tool used for patient involvement in the NG), social media channels and mailings from Freya, the Dutch Patients’ Association for infertility” [51]
	Demonstrated proficiency in technical methods	“Using web-based online voting software, the members of the expert panel, including patient representatives, were invited to participate in three online voting and commenting rounds” [38]
Growth in patient engagement	Patient calls for patient engagement	“Patient groups themselves have called for ‘greater public and patient involvement’ in NICE’s work. The Multiple Sclerosis (MS) Society has asked NICE to go further and fund independent research into barriers to involvement and how to overcome them; fund training and workshops on the reality, not just the theory, of being involved with a NICE appraisal; and provide best practice examples of effective patient organization submissions” [30]
	Professionals’ calls for patient engagement	“In medicine, there are increasing calls for a move toward decision-making that is more inclusive and democratic, reflecting the notion that people should be involved in their own governance” [43]
	Patient engagement as goal in itself	“Considering patients’ preferences is interpreted in terms of active involvement and influence over CPG development and clinical decision-making. Fostering greater patient influence is seen as a goal in itself, justified by the role of patients as beneficiaries of care. Active methods of patient involvement (ex. patient representation in CPG development group), are favored over passive consultation methods... . Ultimately, genuine consumer involvement in CPG development is assumed to lead to greater empowerment of individual consumers, greater access to beneficial treatments, and better self-management” [34]

CPG Clinical Practice Guideline, CEO Chief Executive Officer, NICE National Institute for Health and Care Excellence, COPD chronic obstructive pulmonary disease, TBI traumatic brain injury, NG Network Guideline

patient perspective beyond the categorical demographic data typically captured in online methods [37]. Examples include disentangling analytically-deduced views from personal narratives that are overly passionate or influenced by cognitive dissonance [42, 45], as well as potential inaccuracies in estimating patient preferences by caregivers and the public [35] or professional patients [31] as proxies. The added barrier of *remote* engagement could further complicate accounting for these complexities of patient engagement. Furthermore, online methods may be biased towards including younger patients [46], patients with more education or without cognitive impairment [47], patients with stable Internet access [48], and patients with chronic conditions [49]. However, these challenges are increasingly mitigated by growing demands for patient engagement by both patients [30] and professionals [43]

who recognize patient engagement as a goal itself [34], as well as patients’ increasing familiarity with using online technologies for both CPG and overall healthcare activities [38, 50, 51].

4 Discussion

Based on our rapid review and qualitative evidence synthesis, we identified several themes capturing possible advantages and challenges of online patient engagement methods. Advantages include the greater openness anonymity can facilitate, potential to improve the utility of CPG products, and the ability to reach more patients. Challenges of online methods include the difficulty of ensuring patients understand their tasks, the need to account for

diversity in patient perspectives, time and resources required, and less engaging nature of online methods compared with in-person methods, although these challenges can be mitigated by patients' preference for being engaged and increasing patient fluency with online methods. While these advantages, challenges, and mitigating factors may also apply in varying degrees to in-person methods, our findings suggest that online engagement of patients in CPG development is most likely to be useful when patients seek anonymity to feel comfortable to share their views openly; a large, diverse, and geographically dispersed group of patients is sought; patients have access to and proficiency with online technology; and the GDG has time and resources to train patients, ensure they understand their tasks, and manage the needs of special patient populations.

Our process of inferring analytic themes from descriptive themes, which were developed from line-by-line codes of primary studies, aims to summarize our findings in a way that is conceptually rich yet empirically faithful to data from the primary studies [29], rather than producing entirely speculative inferences. However, several limitations should be noted when interpreting our findings. First, rapid reviews do not incorporate all of the rigorous components of systematic reviews to make allowances for the rapid synthesis of identified information [20, 21]. We limited our search to a select number of major databases as well as specific articles identified by our team and advisory board members, leading to the possibility of missing some published articles or unpublished work in progress discussing online engagement of patients during CPG development [29]. Consequently, it is possible that we did not identify other advantages, challenges, and mitigating factors because they may not have been mentioned in the published literature we reviewed. Moreover, as is common in rapid reviews [21], only one member of our review team screened titles and abstracts, assessed full-texts for eligibility, extracted information from included studies, and synthesized findings. To increase the rigor of our rapid review, we actively engaged our team and advisory board members during the design of all data abstraction and coding activities, and the interpretation of study findings [52]. Lastly, we did not incorporate quality assessments into our review process due to the heterogeneity of sources included and the unique needs of our project.

We also note several areas of future research. First, we identified a paucity of research literature on engaging pediatric patients and their caregivers. Future research should explore the implications of engaging children and using their caregivers as proxies in CPG development. Second, more work is needed on different online methods to engage a large number of patients in CPG development across CPG stages. While involvement of individual

patients is increasingly commonplace, this typically consists of only one or two patients on a guideline panel at the stage when the guidelines are being drafted, or involvement of a larger number of patients once a draft guideline has been posted online for public comment. Future projects should use a framework that includes different levels (e.g. communication, consultation, and participation [48]) and stages of patient involvement in guideline development to systematically examine the advantages and challenges identified in this review to assist GDGs in choosing a specific online patient engagement method. For instance, while our project primarily focuses on patient consultation, it could be explored in participatory and communicative mechanisms. In addition, there is a paucity of evaluations on the comparative utility of different methods for engaging patients in CPG development [7]. Future research is needed to identify best practices among the increasing number of options for patient engagement in CPG development. Moreover, as technology changes, it will be important to continue to assess preferences and utility of online engagement for CPG. Lastly, future research should examine the impact of online methods of patient engagement for different approaches to developing guidelines, such as the RAND/UCLA Appropriateness Method [18] versus the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach [53].

5 Conclusions

GDGs are increasingly involving patients in the development of CPGs. We examined the potential advantages and challenges of using online methods to engage patients in this context. Potential advantages include the ability of online methods to facilitate greater openness and honesty by patients and reflect a greater diversity of patient views. Potential challenges include the extra skill, time, and certain types of resources required. Online methods of engagement are most likely to be useful when patients seek anonymity and have proficiency with online technology, as well as when GDGs desire a large group of patients and have the required resources.

Our group's focus is integrating patients and caregivers into patient-centered guideline development in the context of Duchenne muscular dystrophy. Although members of the research team have successfully engaged the patient and caregiver community for complex issues [15–17, 54–58], the challenges of incorporating patients and caregivers into CPG development are notable. The results from this review will inform our efforts to integrate the perspectives of both parent caregivers and adult patients; a rare disease population that is geographically dispersed; a group of patients who have physical and

mobility limitations that make travel difficult or impossible; and a group of individuals managing a burdensome, time-consuming disorder.

Author contributions SG and DK developed the initial protocol for this study. All authors contributed to the finalization of the protocol. SG examined citations for eligibility, conducted the data extraction, and developed the initial evidence synthesis. All authors reviewed the initial synthesis and revised iteratively. SG and DK wrote the initial draft of this article. All authors contributed to the article and approved the final revised version.

Compliance with Ethical Standards

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Disclaimer All statements in this article, including its findings and conclusions, are solely those of the authors and do not necessarily represent the views of PCORI, its Board of Governors or Methodology Committee.

Conflict of interest Sean Grant's spouse is a salaried employee of Eli Lilly and Company and owns stock in the company; he has accompanied his spouse on company-sponsored travel. Sean Grant and Dmitry Khodyakov are members of the ExpertLens team. Glen S. Hazlewood, Holly L. Peay, Ann Lucas, Ian Coulter, and Arlene Fink declare that they have no conflicts of interest.

Ethical approval This study does not include human subjects and thus ethics approval was not required.

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